Journal of Forensic and Legal Medicine 20 (2013) 395–398

Contents lists available at SciVerse ScienceDirect

Journal of Forensic and Legal Medicine

journal homepage: www.elsevier.com/locate/jflm

Case review

Fatal aortobronchial fistula

Mo Li Medical Student [a,b], Neil Langlois M.D., Forensic Pathologist [a,b], Roger W. Byard M.D., Forensic Pathologist [a,b,*]

[a] The University of Adelaide Medical School, Frome Road, Adelaide 5005, SA, Australia
[b] Forensic Science SA, 21 Divett Place, Adelaide 5000, SA, Australia

ARTICLE INFO

Article history:
Received 20 June 2012
Accepted 27 July 2012
Available online 22 August 2012

Keywords:
Aortobronchial fistula
Haemorrhage
Aneurysm
Surgery
Aorta
Aortitis

ABSTRACT

Aortobronchial fistula is a rare condition characterized by the development of a communication between the aorta and a branch of the bronchial tree that results from processes that arise within the aorta, in the tissues of the mediastinum, or in the adjacent thoracic organs. Three cases are reported to demonstrate characteristic features. *Case 1*: An 82-year-old woman was found collapsed with blood clot in her mouth. At autopsy an atherosclerotic thoracic aortic aneurysm was found which had eroded into the underlying left main bronchus with filling of the larynx, trachea and main bronchi with fluid blood. There was no evidence of dissection. *Case 2*: A 30-year-old woman collapsed and died. At autopsy, coarctation of the thoracic aorta was found with a saccular aneurysm distal to this which was joined to the left main bronchus by a mass of necrotic tissue. The bronchus contained a cast of blood; blood was also present within the trachea and within the distal airways of the left lung. *Case 3*: A six-year-old girl collapsed with massive airway haemorrhage following bronchoscopy. At autopsy coarctation of the thoracic aorta was identified with a saccular aneurysm distal to this. A transverse tear of the thinned aneurysm wall communicated with a mass of necrotic friable tissue that extended through the wall of the left main bronchus. Distal airways were filled with fluid blood. All three deaths were due to haemorrhage from aortobronchial fistulas. The pathogenesis of aortobronchial fistulas involves a variety of mechanical, infective and neoplastic processes. Many cases will not be diagnosed until autopsy examination is performed.

1. Introduction

Aortobronchial fistula is a rare condition characterized by the development of a communication between the aorta and a branch of the bronchial tree.[1] Only 71 cases had been reported prior to 1995.[2] The descending thoracic aorta and the major airways of the left lung are most commonly involved due to their close anatomical proximity.[3] The presentation is typically with haemoptysis which may be intermittent and minor, or massive with a mortality rate of 100% if left untreated.[1,2,4] Other symptoms may include cough, dypnoea and chest pain.[5,6] Approximately 30–50% of cases are first diagnosed at autopsy,[1,3] demonstrating the importance of careful dissection of the upper airways and adjacent vessels in cases of fatal haemoptysis. The following study details characteristic features and underlying causes of this uncommon entity.

2. Materials and methods

Case files at The University of Adelaide Discipline of Anatomy and Pathology and Forensic Science SA, Adelaide Australia, were reviewed for illustrative examples of aortobronchial fistula. Case details were summarized. A literature search was also performed of the United States National Library of Medicine 'Entrez PubMed' database (http://www.ncb.nlm.nih.gov/pubmed) for all entries listed under "aortobronchial fistula".

3. Results

Three cases were identified.

Case 1: An 82-year-old woman was found collapsed with blood clot in her mouth. She had been complaining of back and abdominal pain for approximately two weeks but had not sought medical attention. Her past history included hypertension, a known thoracic aortic aneurysm and a left hemicolectomy for carcinoma of the colon. At autopsy the major findings related to an atherosclerotic thoracic aortic aneurysm. There was no evidence of dissection, however, the aneurysm had eroded into the underlying left main

* Corresponding author. Discipline of Anatomy and Pathology, Level 3 Medical School North Building, The University of Adelaide, Frome Road, Adelaide 5005, SA, Australia. Tel.: +61 8 8226 7700; fax: +61 98 8226 7777.
E-mail address: roger.byard@sa.gov.au (R.W. Byard).

1752-928X/$ – see front matter © 2012 Elsevier Ltd and Faculty of Forensic and Legal Medicine. All rights reserved.
http://dx.doi.org/10.1016/j.jflm.2012.07.008

bronchus with filling of the larynx, trachea and main bronchi with fluid blood. Underlying but incidental disease included marked coronary artery atherosclerosis, a previous hemicolectomy and metastatic adenocarcinoma to the liver. Death was due to haemorrhage from an aortobronchial fistula caused by pressure necrosis from a thoracic aortic aneurysm.

Case 2: A 30-year-old woman awoke with blood on her face and then collapsed and died. At autopsy, coarctation of the aorta was found immediately distal to the origin of the left subclavian artery with a saccular aneurysm beyond this. The aneurysm was joined to the left main bronchus by a mass of necrotic tissue. The bronchus contained a cast of blood; blood was also present within the trachea and had been aspirated into the distal airways of both lobes of the left lung. No other significant findings were present and there was no evidence of any syndromal condition such as Turner syndrome. Death was attributed to haemorrhage from an aortobronchial fistula due to erosion by a post-coarctation aortic aneurysm.

Case 3: A six-year-old girl collapsed with massive airway haemorrhage following bronchoscopy for obstructive respiratory symptoms. It is not clear whether she had suffered from haemoptysis. At autopsy coarctation of the aorta was identified immediately beyond the origin of the left subclavian artery where the luminal diameter narrowed to 5 mm. Distal to this a saccular aneurysm (diameter 2 cm) was pressed firmly into the hilar region of the lung against the left main bronchus. A transverse tear of the thinned aneurysm wall communicated with a mass of necrotic friable tissue that extended through the wall of the left main bronchus approximately 2 cm from the carina (Figs. 1 and 2). The necrotic material extended into the bronchial lumen with narrowing. Distal airways were filled with fluid blood. The aortic valve was bicuspid with no other significant findings or syndromal features present. Death was attributed to haemorrhage from an aortobronchial fistula (caused by a post-stenotic aortic aneurysm) following bronchoscopy.

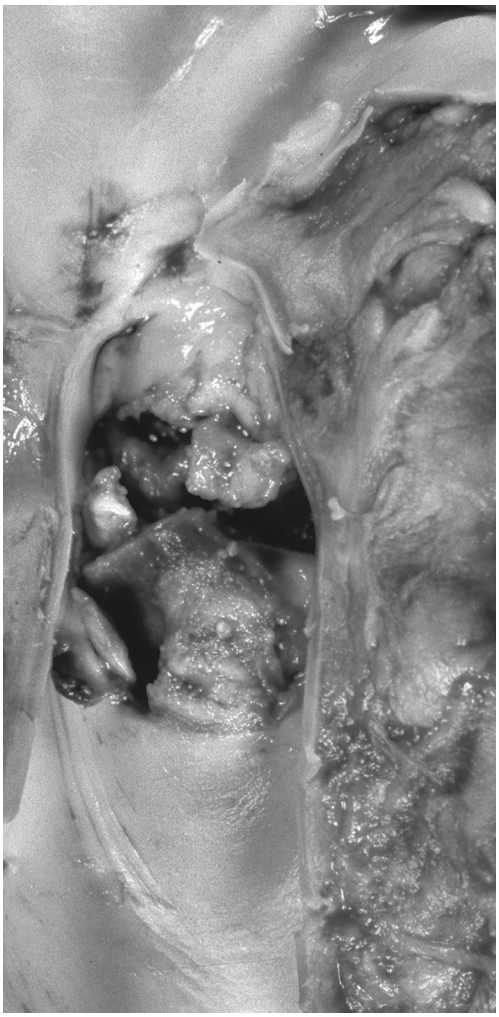

Fig. 1. Tearing of the wall of a post-stenotic saccular aortic aneurysm in a six-year-old girl with exposure of underlying necrotic tissues (case 3).

4. Discussion

Fistulas forming between the aorta and the oesophagus or bronchi may have similar presentations with herald haemorrhages producing expectoration of blood,[7] followed by exsanguination. Distinguishing between these two entities may only be possible at autopsy. Intermittent haemoptysis may occur if blood clot sealing off the fistula tract lyses or is dislodged.[2] As the reported cases have demonstrated, aortobronchial fistulas may arise at any age.

Primary aortic pathology initiating fistulas most often involves aneurysms from a variety of aetiological causes (Table 1). As aneurysms expand they exert pressure on the adjacent tracheobronchial tree with tissue necrosis and eventual breakdown of the intervening vascular and bronchial walls, allowing direct communication between the circulation and the lumen of the upper airways. Aortic atherosclerosis is one of the more common conditions leading to aneurysm formation with erosion into the airways/lungs causing fatal haemoptysis,[8] as in case 1. Other types of aortic aneurysms may also cause aortobronchial fistulas by a similar

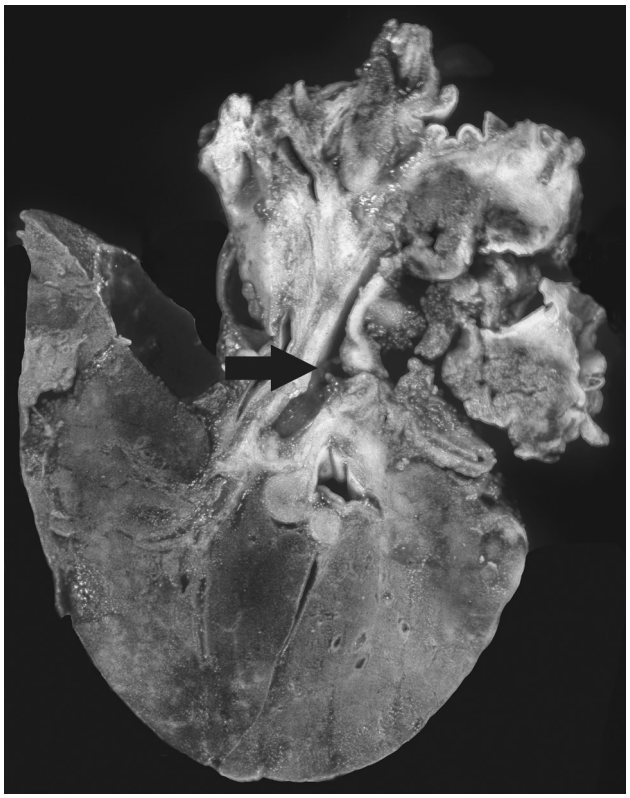

Fig. 2. The other side of the aortobronchial fistula in Fig. 1 with ulceration of the bronchus overlying a defect communicating with the aorta (arrow). It appears most likely that the bronchoscope dislodged adherent clot/necrotic tissue that had temporarily sealed the defect.

Table 1
Causes of lethal aortobronchial fistula.

Primary aortic involvement
i) Aneurysm
a) Atherosclerotic
b) Infective/inflammatory
c) Post-traumatic
ii) Trauma
a) Sharp/blunt chest injury
b) Penetrating ingested/inhaled foreign body
iii) Post-surgical
Secondary aortic involvement
i) Malignancy
a) Pulmonary
b) Mediastinal
ii) Infection
iii) Iatrogenic
a) Tracheostomy
b) Bronchoscopy
c) Post-surgical

mechanism of pressure necrosis, as was observed in cases 2 and 3 with post-coarctation aortic dilation. The association of aortic coarctation and dilation with a bicuspid aortic valve[9] was also demonstrated in case 3. Fistulas may complicate aortic dissection without the presence of an aneurysm.[3,10]

Mycotic aneurysms may arise in the setting of infective aortitis or endocarditis[10,11] with infection causing inflammation, necrosis, and weakening of the aortic wall leading to aneurysm formation. Non-infectious Inflammatory conditions such as giant-cell arteritis can also have a similar effect.[12]

Direct injury to the aorta from blunt or sharp chest trauma may result in the development of asymptomatic aneurysms which expand over time and press against the tracheobronchial tree leading to fistulas.[3] Penetrating sharp trauma to the chest such as a stab wound may also create an acute aortobronchial fistula, however the presentation is usually marked by pulmonary collapse and filling of the pleural space with blood. An alternative form of traumatic fistula may result from migrating ingested foreign bodies.[13]

Aortic surgery may also lead to fistula formation at variable times after surgery. Surgery may be for congenital defects such as a patent ductus arteriosus or coarctation in infancy, for repair of a thoracic aortic aneurysm, or more rarely for resection of a primary aortic tumour such as a sarcoma, in later life.[14] Aortobronchial fistulas may also be caused by failure of an anastomosis in a graft replacement or a patch aortoplasty.[2,15] Fistulas are believed to result from pseudoaneurysms which develop following disruption of the aortic wall with haematoma formation.[14] This process may be exacerbated by inappropriately tight suturing, foreign material, infection of prosthetic grafts and host factors such as fragility of the aortic wall or atherosclerosis.[14]

The wall of the aorta may be breached by pathological processes arising in the adjacent lung or soft tissues. For example, primary bronchogenic carcinomas may infiltrate the aorta,[16] as may secondary neoplastic lesions of the para-aortic lymph nodes. On occasion the aorta may be literally encased by a mass of malignant tissue.[17]

Since the first description in 1914 of an aortobronchial fistula in a patient with tuberculosis there have been numerous reports detailing other causes, including infection.[2,14] As noted above, infections may result in expanding mycotic aneurysms, but may also directly initiate fistula formation. Infectious agents may spread to the aorta via the circulation, by direct contamination during surgery or from nearby pneumonia.[3] A common bacterial pathogen is Staphylococcus with less common infections involving syphilis and mycobacterium tuberculosis.[3,5,18] Tuberculous infections may arise in the lungs or within pulmonary hilar lymph nodes leading to extensive local caseous necrosis with involvement of the aorta, bronchi or oesophagus.[7,19] Although it has been suggested that invasive pulmonary aspergillosis may directly infiltrate into the aorta and cause massive haemoptysis due to the formation of an aortobronchial,[20] fistula, the possibility of fungal embolization into thoracic branches of the aorta as the initiating process should be considered.[21] Pulmonary mucormycosis is another fungal condition that may create a fistula between the aorta and the lung[22] as, very rarely, may hydatid disease.[23]

Iatrogenic complications that may lead to an aortobronchial fistula were demonstrated in case 3 where lethal haemoptysis followed bronchoscopy, most likely due to disruption of a mass of necrotic tissue that had eroded the wall of the bronchus, narrowing the lumen. Bronchoscopy may also dislodge adherent blood clot that has sealed the defect leading to fatal haemoptysis.[2] Tracheostomy has been related to the formation of a fistula between major arteries and the trachea. The mechanisms include direct penetration by the tip of the tracheostomy tube or pressure necrosis, and usually involve branches of the aorta, rather than the aorta itself. Prolonged intubation and a low tracheal incision are predisposing factors to tracheovascular fistulas.[24,25] Aortobronchial fistulas from eroding metal stents have been reported in children months to years after treatment of bronchomalacia,[2] and also rarely after oesophagectomy in an adult from pressure necrosis from eroding vascular clips.[26]

This overview was undertaken to demonstrate an uncommon event that may have devastating consequences. The pathogenesis of aortobronchial fistulas is diverse involving mechanical, infective and neoplastic processes. Given the speed at which lethal exsanguinations may occur, the diagnosis may not be established until the time of autopsy.

Ethical approval
Forensic Science South Australia.

Funding
None.

Conflict of interest
None.

References

1. Falode T, Alphonso N, Austin C, Anderson D. Aortobronchial fistula: the hidden connection. *Asian Cardiovasc Thorac Ann* 2006;**14**:e74–5.
2. Oppenheimer R, Brotherton L. Aortobronchial fistula: a rare etiology for hemoptysis. *Ear Nose Throat J* 2002;**81**:257–9.
3. Fernández-González AL, Montero JA, Luna D, Gil O, Sanjuán VM, Monzonis AM. Aortobronchial fistula secondary to chronic post-traumatic thoracic aneurysm. *Tex Heart Inst J* 1996;**23**:174–7.
4. Demeter SL, Cordasco EM. Aortobronchial fistula: keys to successful management. *Angiology* 1980;**31**:431–5.
5. MacIntosh EL, Parrott JCW, Unruh HW. Fistulas between the aorta and tracheobronchial tree. *Ann Thorac Surg* 1991;**51**:515–9.
6. Yoo J-H, Lee C-T, Shim Y-S, Chung JW, Ahn H, Kim KW. Aortobronchial fistula presenting as recurrent hemoptysis and successfully treated with an endovascular stent graft. *Respiration* 2001;**68**:537–9.
7. Byard RW. Lethal aortoesophageal fistula – characteristic features and etiology. *J Forensic Leg Med* 2013;**20**:164–8.
8. Tsokos M, Byard RW. Massive, fatal aspiration of blood: not necessarily a result of trauma. *Am J Forensic Med Pathol* 2007;**28**:53–4.
9. Yuan S-M, Jing H. The bicuspid aortic valve and related disorders. *Sao Paulo Med J* 2010;**128**:296–301.
10. Riancho JA, Gutiérrez JA, Echevarría S, Daroca R, González Macías J. Aortobronchial fistula: an uncommon cause of haemoptysis. *Postgrad Med J* 1989;**65**:171–3.
11. Byramji A, Gilbert JD, Byard RW. Sudden death as a complication of bacterial endocarditis. *Am J Forensic Med Pathol* 2011;**32**:140–2.
12. Caspary L. Vasculitides of large vessels. *Vasa* 2011;**40**:89–98.

13. Taha AS, Nakshabendi I, Russell RI. Vocal cord paralysis and oesophago-broncho-aortic fistula complicating foreign body-induced oesophageal perforation. *Postgrad Med J* 1992;**68**:277–8.
14. Picichè M, De Paulis R, Fabbri A, Chiariello L. Postoperative aortic fistulas into the airways: etiology, pathogenesis, presentation, diagnosis, and management. *Ann Thorac Surg* 2003;**75**:1998–2006.
15. Kazerooni EA, Williams DM, Abrams GD, Deeb GM, Weg JG. Aortobronchial fistula 13 years following repair of aortic transaction. *Chest* 1994;**105**:1590–4.
16. Mosavy SH, Ashrafi MA, Vakhshuri P. Death due to gross vascular perforation by pulmonary carcinoma. *Angiology* 1977;**28**:48–51.
17. James CL, Byard RW, Knight WB, Rice MS. Metastatic osteogenic sarcoma to the heart presenting as bacterial endocarditis. *Pathology* 1993;**25**:190–2.
18. Lee EB, Lee SC, Cho JY, Lee JT. Surgery for concomitant aortoesophageal and aortobronchial fistula in tuberculous aortitis. *Int Cardiovasc Thorac Surg* 2003;**2**:234–6.
19. Masjedi M-R, Davoodian P, Forouzesh M, Abttahi SJ. Broncho-aortic fistula secondary to pulmonary tuberculosis. *Chest* 1988;**94**:199–200.
20. Islam S, Williams DM, Teitelbaum DH. Aortobronchial fistula from invasive *Aspergillus* infection of the lung: an endovascular approach to repair. *J Pediatr Surg* 2005;**40**:E19–22.
21. Byard RW, Jimenez CL, Carpenter BF, Hsu E. Aspergillus-related aortic thrombosis. *Can Med Assoc J* 1987;**136**:155–6.
22. Ohguchi H, Sai T, Hamazaki Y, Kameoka J, Sasaki T. Aortobronchial fistula induced by pulmonary mucormycosis in acute myeloid leukemia. *Rinsho Ketsueki* 2004;**45**:387–92.
23. Buchholz S, Sowden D, Stapleton T, Pohlner P, Wright C. Massive haemoptysis due to aortobronchial fistula caused by pulmonary hydatidosis. *Med J Aust* 2009;**190**:222.
24. Byard RW, Gilbert JD. Potentially lethal complications of tracheostomy: autopsy considerations. *Am J Forensic Med Pathol* 2011;**32**:352–4.
25. McCormick B, Manara AR. Mortality from percutaneous dilatational tracheostomy. A report of three cases. *Anaesthesia* 2005 May;**60**:490–5.
26. Li H-P, Hsieh C-C, Chiang H-H, Wang T-H, Lee J-Y, Huang M-F, et al. Aortobronchila fistula afte esophagectomy for esophagela cnacer – a very rare complication. *Kaoh J Med Sci* 2011;**27**:247–50.